Formosan Journal of Surgery (2015) 48, 65–67

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CASE REPORT

Invasive ductal carcinoma arising in borderline phyllodes tumor

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Received 8 May 2014; received in revised form 10 July 2014; accepted 1 October 2014
Available online 7 May 2015

KEYWORDS
invasive ductal carcinoma;
mammography;
phyllodes tumor;
ultrasonography

Summary Phyllodes tumors (PTs) are an infrequent breast tumor presentation. An invasive carcinoma arising in a borderline PT is extremely rare. A 58-year-old female presented with a mass over the left breast, which she had had for >1 year, with rapid growth in the past 3 months. The patient underwent wide excision under the assumption that the mass was a PT. Invasive ductal carcinoma and ductal carcinoma *in situ* arising in a borderline PT were incidentally found. Because the surgical margin was <1 mm, the patient underwent a partial mastectomy to provide a safe margin and a sentinel node biopsy. The pathology of the second excision revealed neither a residual tumor nor lymph node metastasis. The pathologic stage was T2N0M0. This was an extremely rare case of invasive ductal carcinoma arising in a borderline PT, and the clinical course, image, and histological findings are discussed in this paper.

1. Introduction

The term *phyllodes* is derived from the Greek word *phyllon* (meaning leaf) and refers to a rapidly growing, large breast tumor that can cause breast distortion and pressure necrosis of the overlying skin. The tumor appears leaflike in cross section and originates from the periductal stromal cells of the breast. Such tumors account for <1% of all breast neoplasms. Invasive carcinoma arising in malignant phyllodes tumors (PTs) has been reported in previous studies.[1,2] In this paper, we present a rare case with the initial impression of PT. The final diagnosis was invasive ductal carcinoma and ductal carcinoma *in situ* arising in a borderline PT.

Conflicts of interest: The authors have no conflicts of interest.

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http://dx.doi.org/10.1016/j.fjs.2014.10.002

2. Case report

A 58-year-old female patient had a palpable mass over the left breast for >1 year, but she did not undergo any examination. She came to our hospital because the tumor had grown rapidly in the past 3 months. She has no family history of breast cancer. A physical examination revealed a well-defined, freely movable, and elastic mass 7 cm × 5 cm in size, located in the 3 o'clock direction and 3 cm from the left breast nipple. There were no palpable lymph nodes over the supraclavicle or axillary region.

A mammography showed a high-density mass approximately 6.6 cm in length with an obscured margin over the upper outer quadrant of the left breast (Fig. 1). An ultrasonography revealed a well-defined, lobulated, and hypoechoic mass, approximately 6 cm × 4 cm in size, in the left breast without enlarged lymph nodes over the bilateral axillary region (Fig. 2).

Tumor markers, such as CEA and CA15-3, were within normal limits. Fine-needle aspiration cytology revealed clusters of benign, proliferative ductal epithelia and bipolar cells but no malignant cells. Under the impression that the patient had a PT of the left breast, a wide excision was performed, and a well-defined, soft-to-firm, rubber-like nodule 6 cm × 4 cm × 3 cm in size was found.

A histological study revealed a well-circumscribed tumor with a focal infiltrative border composed predominantly of fibroadenoma-like areas focally arranged in a leaflike architectural pattern created by extensive branching of the epithelial component. In addition, stromal hypercellularity with spindle-cell nuclei was found (Fig. 3). The mitotic activity was increased with moderate cellular pleomorphism. The ductal epithelia exhibited a variable degree of hyperplasia. Several ducts were arranged as invasive islands, often angulated, in which well-defined spaces were formed by arches of cells (a sieve-like or cribriform pattern). Part of the tumor was composed of a single layer of epithelial cells enclosing a clear lumen. These tubules were generally ovate or rounded. The tumor cells were small and regular with little evidence of nuclear pleomorphism. Mitoses were clearly observed in several areas. In addition, a prominent reactive-appearing fibroblastic stroma was present.

Figure 1 Mediolateral oblique mammography showed a high-density mass with an obscured margin over the upper outer quadrant of the left breast.

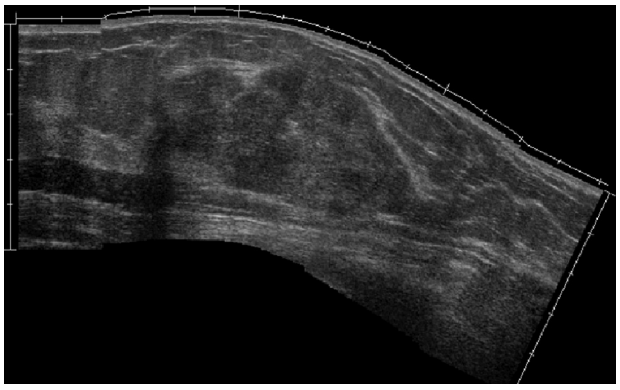

Figure 2 Ultrasound examination of the left breast revealed a well-defined, lobulated, and hypoechoic mass measuring 6 cm × 4 cm.

The formal pathology report stated that the tumor consisted of Grade 1 invasive ductal carcinoma and cribriform type, intermediate grade, borderline *in situ* ductal carcinoma arising in a PT, with the largest invasive carcinoma being 2.5 cm × 1 cm in size. The carcinoma component stained 99% for estrogen receptor, 90% for progesterone receptor, and 1+ for HER2/neu (Fig. 4). The absence of basal myoepithelial lining was confirmed by negativity for p63. The surgical margin was <1 mm from the PT.

Two weeks later, the patient underwent a partial mastectomy to provide a safe margin and a sentinel node biopsy. The pathology of re-excision showed neither a residual tumor nor lymph node metastasis. The pathologic stage was T2N0M0. The patient was discharged and had an uneventful postoperative course.

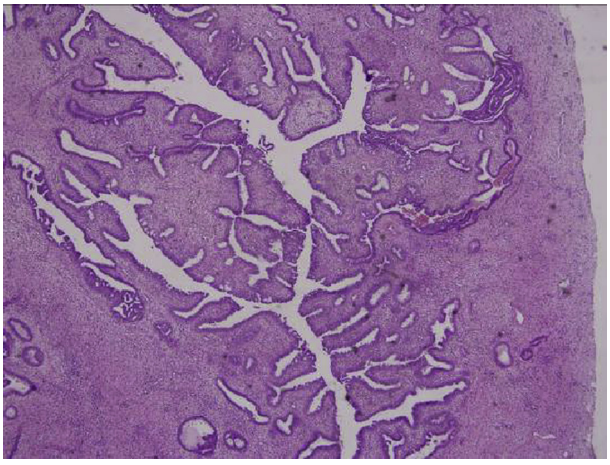

Figure 3 Leaflike structures of the epithelial component with stromal hypercellularity (×40).

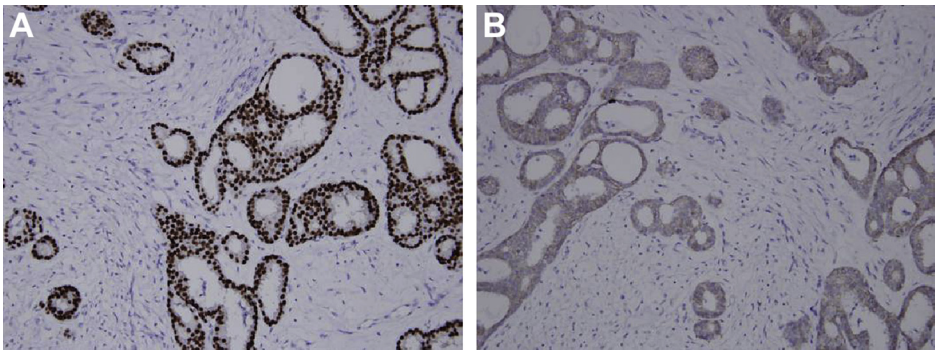

Figure 4 The carcinoma component stained (A) 99% for the estrogen receptor (immunohistochemistry, ×200); (B) 1+ for HER2/neu (immunohistochemistry, ×200).

3. Discussion

PTs are fibroepithelial tumors composed of epithelial and cellular stromal components. They were first described by Johannes Mueller in 1838.[3] Histologically, they may be considered benign, borderline, or malignant, depending on certain features including degree of stromal cellular atypia, type of tumor margin (infiltrative vs. circumscribed), mitotic activity, and the presence or absence of stromal overgrowth.[4,5]

PTs are typically benign and can be cured through wide excision with histologic margins negative for malignant cells.[6] A histologic margin of at least 1 cm has been reported in previous studies.[4,7] The stromal component may undergo malignant transformation; hence, the misleading traditional term *cystosarcoma phyllodes*. However, it is rare for epithelia to undergo such a change, which usually occurs in cases of malignant PTs.[1,2]

The patient in this study exhibited a rapidly enlarging left breast mass corresponding to the clinical course of PTs. The mammography revealed a high-density mass with an obscured margin and the sonography revealed a well-defined, lobulated, and hypoechoic mass, which constitute typical findings of a PT.[8] However, PTs are difficult to distinguish from other tumors on the basis of clinical history and radiologic features.[9]

Treatment of these rare cases should be tailored to address the PTs and invasive carcinomas individually. PTs are treated through wide excision, and invasive carcinomas should be treated using a method including sampling/dissection of the axillary lymph nodes as well as adjuvant therapy based on the pathologic stage, tumor estrogen receptor, progesterone receptor, and HER2/neu status. Lenhard et al[10] reported the local recurrence of borderline PT to be 20%. The prognosis of invasive ductal carcinoma is based on the classification of conventional histology and immunohistochemistry. Currently, there is little consensus on the prognosis of this rare disease.

In conclusion, surgical resection of the tumor with a safe margin and thorough sampling of the tumor are necessary to fully assess its biologic characteristics and detect areas of stromal overgrowth that suggest malignant behavior. In addition, it is crucial to be aware that rare cases of carcinoma may arise within a PT.

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